Treatment of pulmonary aspergilloma in cystic fibrosis by percutaneous instillation of amphotericin B via indwelling catheter

P J Ryan, D E Stableforth, J Reynolds, K M Muhdi

Abstract

Pulmonary aspergilloma is a rare complication of cystic fibrosis and is a contraindication to transplantation. The elimination of an aspergilloma in a 24 year old patient with cystic fibrosis by percutaneous instillation of amphotericin B is described, enabling her to be accepted on a lung transplantation programme.

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Keywords: cystic fibrosis, aspergilloma, *Aspergillus*, amphotericin B.

Case report

This patient first came under our care in March 1991 at the age of 20 having been diagnosed as having cystic fibrosis at the age of two years following recurrent pneumonia. In January 1990 she had a right sided pneumothorax treated by intercostal drainage. At the time of referral she weighed 75% of ideal body weight. There was no haemoptysis, but she did have occasional right sided pleuritic chest pain. Her forced expiratory volume in one second (FEV_1) was 0·7 l and forced vital capacity (FVC) was 1·7 l (21% and 41% of predicted). She required pancreatic enzyme supplements and had not been taking inhaled or oral steroids. There was no history of diabetes or tuberculosis. She regularly grew *Pseudomonas aeruginosa, Staphylococcus aureus,* and *Aspergillus fumigatus* in her sputum. Serum *Aspergillus* precipitins were positive and her skin prick test to *Aspergillus* showed no immediate or delayed reaction. Her chest radiograph showed typical features of long standing cystic fibrosis with overinflated lungs and widespread peribronchial thickening. In addition, a cavity was seen in the right upper lobe. A thoracic computed tomographic (CT) scan showed that the cavity contained an aspergilloma adjacent to the visceral pleura (fig 1a). The cavity was entered at fibreoptic bronchoscopy where a grey mass of tissue was seen partially adherent to the wall. A biopsy specimen of the tissue revealed granulation tissue, inflammatory cells, and septate fungi. Treatment with oral itraconazole was started.

Poor pulmonary function led to referral for lung transplantation but she was thought unsuitable because of likely intraoperative contamination of the pleural space and airways by *Aspergillus*. We considered whether the aspergilloma might be excised before transplantation but the opinion of the thoracic surgeons was that the dangers of pleural dissemination, infection and respiratory failure would be too great. A one month course of intravenous amphotericin (1 mg/kg/day) failed to show any evidence of a change in the aspergilloma on the CT scan.

Following reports of successful intracavitary treatment of aspergillomas by an indwelling catheter it was decided to attempt this method.[1,2] A 6·3 French polyethylene pigtail catheter (William Cook Europe, Bjaeverskov, Denmark) was inserted into the cavity using local anaesthesia and CT scan guidance (fig 1b). An initial dose of 5 mg amphotericin B was injected, gradually increasing to 50 mg/day in 20 ml 5% dextrose. Later a smaller total volume of 5 ml was found to be best. A total of 3 g was given over 60 days. Initially the instillation of this solution caused coughing which was relieved by the instillation of 2–4 ml of 1% lignocaine into the cavity before the amphotericin B. The method was taught to the

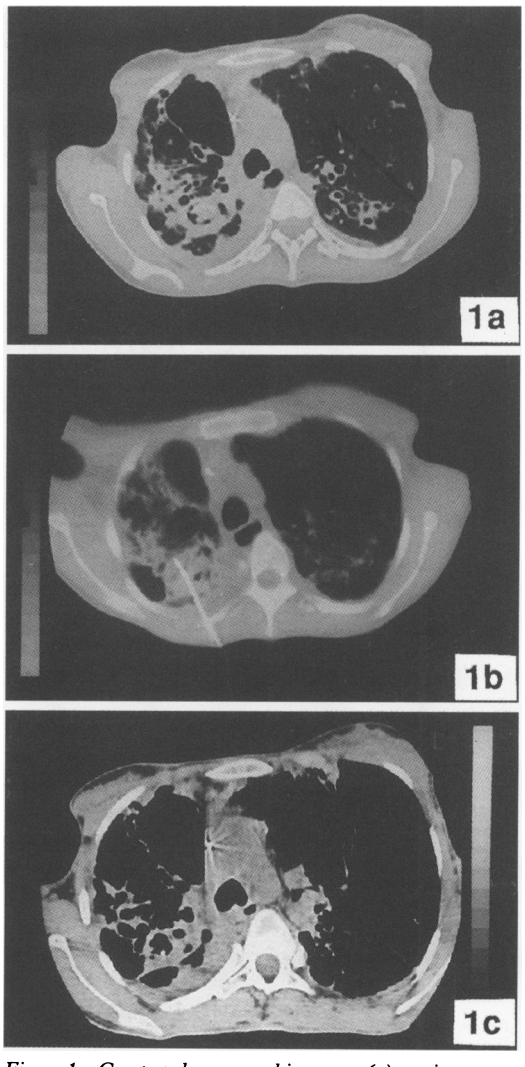

Figure 1 Computed tomographic scans: (a) cavity containing an aspergilloma before treatment; (b) percutaneous catheter lying in the aspergilloma; (c) empty cavity at the end of treatment with intracavitary amphotericin B.

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patient so that most of the course was received at home.

A CT scan of the chest taken at the end of treatment shows an empty cavity (fig 1c). The cavity has remained empty for four months and she continues to take oral itraconazole as her sputum still grows *Aspergillus*. She has been referred back to a transplant centre and has been accepted as a suitable candidate for the active transplant list.

Discussion

Aspergilloma of the lung grows in a pre-existing cavity and usually requires treatment if the patient develops symptoms, most commonly severe haemoptysis. The cavity may communicate with a bronchus and is occasionally viewed directly by fibreoptic bronchoscopy.[3] Medical treatment with either oral antifungal agents or intravenous amphotericin B has a poor record of success.[4,5] Surgery by lobectomy or marsupialisation may be required to clear an aspergilloma, but because of underlying lung disease it carries a high morbidity and mortality.[5,6]

We report the successful clearance of an aspergilloma in a patient with cystic fibrosis by percutaneous instillation of amphotericin directly into the cavity. We were encouraged in this approach by the report by Jackson and colleagues of four patients with aspergilloma treated with intracavitary instillation of amphotericin by an indwelling catheter. In one patient the fungal ball cleared completely, resolution of symptoms occurred in two patients, and there was no effect in the fourth.[1] A Korean group treated seven patients with haemoptysis caused by an aspergilloma using an indwelling catheter in four patients with twice daily cavitary injection in three patients. Haemoptysis ceased in all, and in three of the patients the aspergilloma cleared completely.[2]

Patients with cystic fibrosis and aspergilloma are likely to have many cysts and dilated bronchi colonised by *Aspergillus*. This may be a limiting factor in the success of this approach, with recurrence in the same cavity or another, although the treatment could be repeated if necessary. Only two cases of cystic fibrosis with an aspergilloma have been reported, though with longer survival and a higher prevalence of diabetes mellitus it may become more common.[7] We hope that this treatment has led to long term resolution of the aspergilloma and that our patient will now undergo successful lung transplantation.

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Colonisation with *Aspergillus* of an intralobar pulmonary sequestration

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Abstract

Pulmonary sequestration is a term used to describe an area of embryonic lung tissue supplied by an anomalous systemic artery. Two forms are recognised – extralobar and intralobar – with different clinical presentations. A patient is reported with intralobar pulmonary sequestration in the left lung and colonisation with *Aspergillus* which was successfully treated by lower lobectomy.

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Keywords: pulmonary sequestration, *Aspergillus*, aspergilloma.

Pulmonary intralobar sequestration, although uncommon, is seen more frequently than the extralobar type. The sequestered lung shares a common pleural covering with normal pulmonary tissue. The areas most frequently affected are the medial and posterior basal segments of the lower lobes, with the left being involved twice as often as the right. Pulmonary intralobar sequestration does not usually communicate with the normal bronchial tree although this communication can occur following infection. It is characteristic of both intralobar and extralobar sequestration that the blood supply is from a systemic artery usually arising from the aorta and, with extralobar sequestration, most commonly from the abdominal aorta.[1]

We report a case of an unusual presentation of pulmonary intralobar sequestration with colonisation by *Aspergillus*.

Case report

A 28 year old woman with no known history of pulmonary disease presented with fever, cough and sputum. A diagnosis of left lower lobe pneumonia was made and antibiotics were prescribed with improvement except for persistent weakness. A month later she developed further respiratory symptoms associated with fever and was admitted to hospital. There were no con-